CASE REPORT

Keratocystic odontogenic tumor: Treatment modalities: Study of 3 cases

B Roopak, M Singh, A Shah, G Patel

Departments of Oral and Maxillofacial Surgery, Rajarageshwari Dental College and Hospital, Bangalore, Karnataka, India

Abstract

Management of the keratocystic odontogenic tumor has been one of the most controversial entities of the maxillofacial surgery. It can become quite large because of its ability for significant expansion, extension into adjacent tissues and rapid growth and also has high reccurence rate as it shows a thin, friable wall, which is often difficult to enucleate from the bone in toto, and have small satellite cysts within the fibrous wall. There are various treatment modalities suggested from conservative to radical approach, but studies have shown even with conservative approach good results are achieved. In this paper we are presenting various treatment modalities and 3 cases have been discussed.

Key words: Keratocystic odontogenic tumor, odontogenic keratocyst, reccurence

Date of Acceptance: 11-Jun-2013

Introduction

The World Health Organization (WHO) in its recent (2005) classification of Head and Neck Tumors reclassified the odontogenic keratocyst as a benign neoplasm, recommending the term "keratocystic odontogenic tumor" (KCOT). It is defined as "a benign uni- or multicystic, intraosseous tumor of odontogenic origin, with a characteristic lining of parakeratinized stratified squamous epithelium and potential for aggressive, infiltrative behavior." The orthokeratinized variant of the odontogenic keratocyst is not included in the KCOT. WHO "recommends the term keratocystic odontogenic tumor as it better reflects its neoplastic nature."[1,2]

KCOT arises from cell rests of the dental lamina. Approximately, 10% of all jaw cysts. KCOTs have a predilection for males and occur mainly in the second and third decade of life, occurring most commonly in the mandible, mainly in the posterior body, the angle region and the ascending ramus. It extends in the intramedullary space making it difficult to diagnose in the early stage. However, maxillary tumors are more prone to infection, even when small, making it more likely to be diagnosed at an earlier stage of their development. Radiographically, KCOTs may appear as small, round or ovoid, radiolucent lesions with often scalloped, Multilocular and distinct margins. In large cysts, downward displacement of the inferior alveolar nerve has been reported. Histopathologically, KCOT typically shows a thin, friable wall, which is often difficult to enucleate from the bone in toto and have small satellite cysts within the fibrous wall, with columnar epithelium and parakeratinized epithelium. Therefore, odontogenic keratocysts often tend to recur after treatment, which ranges from 13% to 63%.[3,4]

Recommended techniques are enucleation with chemical cauterization (Carnoy's solution), enucleation with (liquid nitrogen) cryotherapy for smaller cyst, marsupialization or decompression followed by secondary enucleation in the large cyst. Bramley recommended radical surgery with resection and bone graft for larger and recurrent cyst, Bataineh advocated resection without continuity defect as standard treatment.

Address for correspondence:
Dr. B Roopak,
Department of Oral and Maxillofacial Surgery, Rajarageshwari Dental College and Hospital, Bangalore - 560 060, Karnataka, India.
E-mail: drbeenaroopak@gmail.com

Access this article online	
Quick Response Code:	Website: www.njcponline.com
	DOI: ***
	PMID: *******

Radical treatment can cause various complications such as facial deformity, loss of teeth, parasthesia. So the conservative management like marsupialization or Marsupalization can be carried out to reduce tumor size and morbidity and followed by enucleation.

Recurring odontogenic keratocysts are usually associated with the nevoid basal cell carcinoma syndrome (NBCCS), in which the recurrence rate is about 82%. 1. The features are skeletal features such as bifid rib, frontal and parietal bossing and mandibular prognathism, together with multiple basal cell carcinomas and palmar and plantar dyskeratosis. Hypertelorism, mental retardation, strabismus, calcification of the falx cerebri and medulloblastoma may also be present. If a patient is having an odontogenic keratocyst that recurs even after repeated surgeries and is associated with parakeratinization, intramural epithelial remnants and satellite cysts, this single recurring cyst can be considered as the only manifestation of NBCCS for the successful management of which the overlying surface epithelium must be excised.[3]

The aim of this study was to discuss the various treatment modalities in the management of KCOT Which helps the surgeon to decide the treatment plan.

Case Reports

Case 1

A 34-year-old male patient reported to us with a pain and swelling on the left side of the face. Patient had undergone extraction of 37 one month back. Examination revealed swelling in the lower jaw on the left side; vestibular sulcus was obliterated and was tender. Ortho pantomogram (OPG) revealed unilocular radiolucency on the left side of mandible extending from root apices of 1st premolar, 2nd premolar and 1st molar till lower border of mandible [Figure 1] pushing inferior alveolar nerve inferiorly. There was no parasthesia.

As the lesion was very large and patient was young, treatment option chosen was decompression and marsupialization to avoid secondary deformity.

Cyst was decompressed by making buccal window biopsy was sent for histopathologic examination and packed with gauze soaked in bismuth, iodoform paste (BIP) [Figure 2]. It was reported as parakeratinized KCOT. Follow-up was carried out for 3 months, once there was obvious reduction in the size of lesion [Figure 3]. Marsupialization was carried out.

Case 2

A 38-year-old male patient reported with a pain and swelling in the lower back tooth on the right side since 2 weeks. Examination revealed swelling on the right side of mandible obliterating vestibular sulcus with tenderness, 48 was missing. OPG revealed multilocular radiolucency extending from distal root apices of 1st molar to angle on the right side [Figure 4] displacing the 3rd molar inferiorly till lower border and inferior alveolar nerve buccally. Occlusal radiograph showed thinned out lingual cortical plate [Figure 5].

Decompression was done for 3 months, once the impacted tooth moved superiorly and bone formation at the lower border was seen, enucleation with excision of overlying mucosa and chemical cauterization was carried out using Carnoy's solution. Histopathology showed the basal layer budding into connective tissue. Mitosis found in the suprabasal layers and parakeratinized corrugated epithelium [Figure 6]. Post operative followup was done for 1 yr, there was good healing intra orally and on OPG [Figures 7 and 8].

Case 3

A 32-year-old female patient reported with a pain in the lower left back region since 1 week. Patient gave the history of OKC which was treated by enucleation and peripheral osteotomy 2 years back. OPG revealed unilocular radiolucency extending from 2nd premolar to 2nd molar region in the edentulous mandible on the left side, involving lower border of mandible [Figure 9]. Occlusal radiograph showed thinning out of lingual cortical plate [Figure 10]. There was no parasthesia. As this was a recurrent lesion and thinning of lingual cortical plate was present, segmental resection was carried out and reconstruction with rib graft and reconstruction plate 2.5 mm titanium was used [Figure 11]. Intra oral and opg view after 2 yrs of followup showing good healing [Figures 12 and 13]. Patient was given removable prosthesis.

Discussion

Some authors advocate a site and size based approach to KCOT treatment planning. "Small keratocysts near the alveolar process a measuring maximum of 1 cm in diameter should be treated by simple excision, but large keratocysts near the base of the skull should be treated by radical excision." this is because of the potential for local invasion of the skull base, which can have catastrophic consequences. Stoelinga's analysis reported recurrences are mainly because of the highly active basal layer of the KCOT epithelial lining if parts of it are left behind.

Case 1

This case was treated initially by decompression and cavity was packed with BIP paste. Follow-up for 3 months was carried out, which showed a gradual decrease in size and good bone formation seen at the lower border.

Marsupialization was first described by Partsch. Decompression, although serving the same function and following same basic principle, are different. They

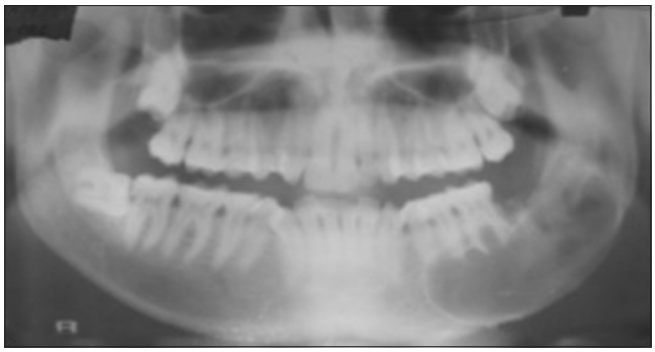

Figure 1: Pre op orthopantomogram showing radiolucency extending from apical region of 34 to ramus region

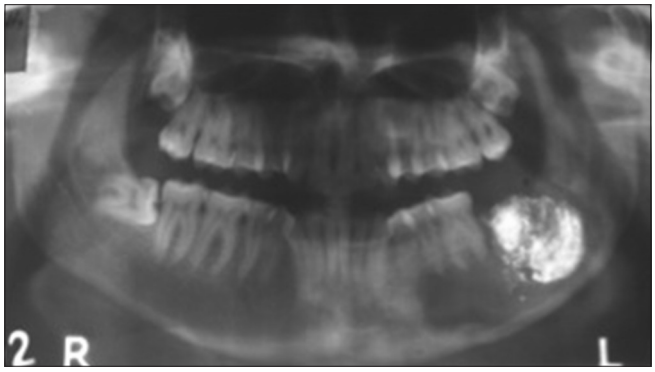

Figure 3: Post op packing with bismuth, iodoform and paraffin paste

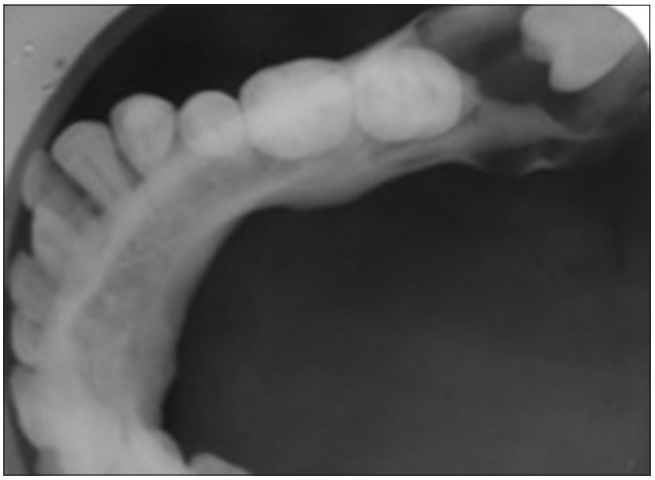

Figure 5: Occlusal X-ray pre op

Figure 2: Intra-oral picture showing bismuth, iodoform and paraffin pack in placed

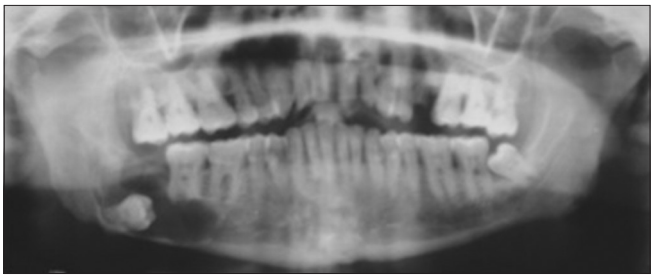

Figure 4: Orthopantomogram pre op showing radiolucency involving implacted 48

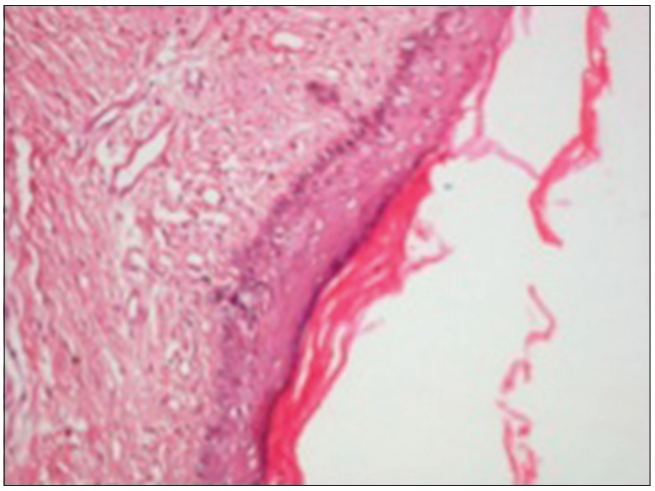

Figure 6: Histopathalogy slide

both relieve the pressure within the cystic cavity, but marsupialization is a one-stage procedure and decompression is a two-stage procedure that requires the placement of a drainage tube, followed by (delayed) enucleation of the residual cyst. Those who criticize this approach argue that marsupialization and decompression does not allow a complete removal of the entire epithelial lining, which might lead to continuous proliferation of the epithelium; thus, facilitating recurrence.[4-6]

Marker *et al*. showed that decompression and marsupialization led to histological changes of the cystic lining, eventually resulting in the replacement by oral epithelium. Presence of inflammation after decompression/marsupialization is thought to change the biologic behavior of the keratocyst into a less aggressive form, with some evidence showing that the epithelial lining transforms into the non-keratinizing epithelium.

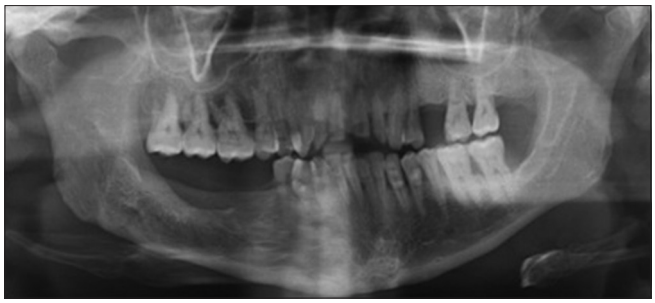

Figure 7: Post op 1 year good healing of bone

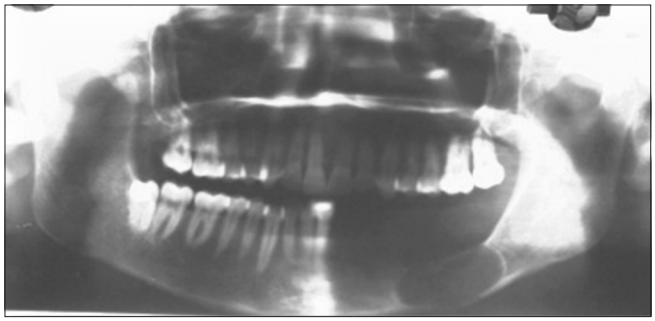

Figure 9: Orthopantomogram showing Unilocular radiolucency extending from 2nd premolar to 2nd molar involving the lower border on the left side of mandible

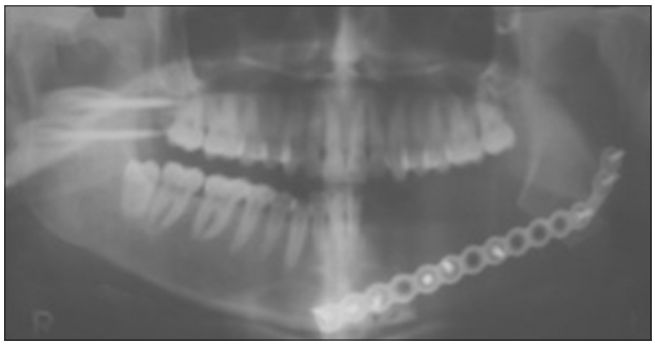

Figure 11: Immediate post op showing rib graft with reconstruction plate

Giuliani *et al*. and Maurette *et al*. stated that this technique required patient compliance for a prolonged period of time, which may be a disadvantage.

Case 2

This case was treated initially by marsupialization for 3 months. Enucleation is carried out within 3 months to avoid risk of phosphate induced bone necrosis. Once there was bone formation at lower border it was treated by Stolinga's method that is enucleation of cyst along with the excision of overlying mucosa and treatment of bony cavity with Carnnoy's Solution.[15,18]

Stolinga suggested Enucleation of the entire lesion along with overlying mucosa, which enables complete removal of lesion with daughter cysts and histopathological examination of the entire lesion. 1st and 2nd and impacted 3rd molar were extracted with lesion to avoid leaving pathologic tissue behind. In general, elimination of the epithelial islands and microcysts located in the overlying, attached mucosa is advised by excising this part of the mucosa.

Figure 8: Intra-oral picture showing post op healing

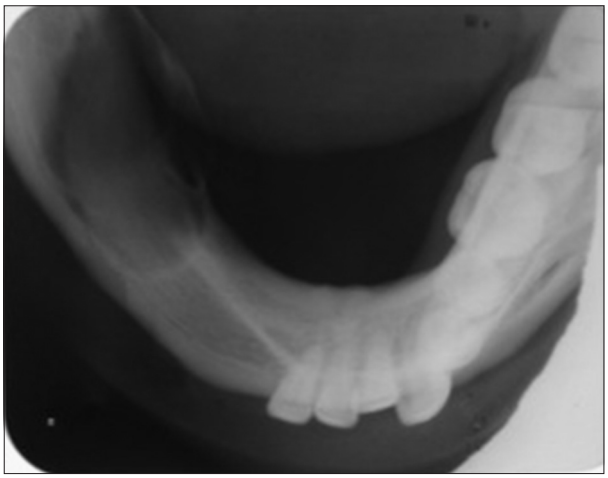

Figure 10: Occlusal X-ray showing unilocular radiolucency and thinning of lingual cortical plate

Figure 12: Intra-oral 1 year follow-up

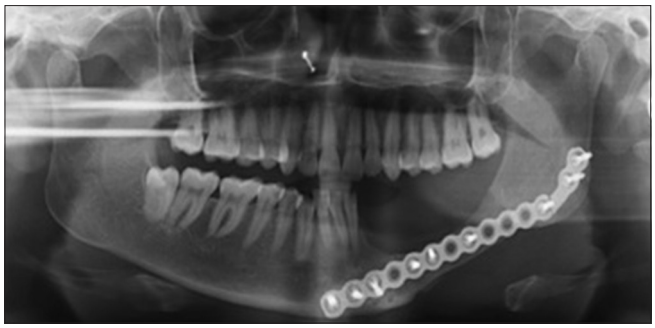

Figure 13: One year follow-up showing healed graft

Chemical cauterization is carried out by using the Carnoy's solution for 3 min to eliminate bony septae, which is placed into bony cavity after enucleation to eliminate epithelial residues from cyst walls that may have been left behind after enucleation thus leading to recurrences.[11,12]

Case 3

Zhao *et al*. reported the most extensive form of treatment indicated for the management of large KCOTs and recurrent lesions is that of osseous resection, marginal or segmental. A mandibulectomy involving approximately 1 cm around the lesion was performed, leaving the lower border of the mandible and the posterior border of the ramus intact. Autogenous grafts or autogenous frozen lesion bone reimplantation was immediately used to reconstruct the defect after segmental mandibulectomy. Although Worrall recommended radical excision as the treatment of choice for KCOTs, which have perforated bone, others consider that resection should be particularly considered for treatment of a recurrent KCOT, in which case it should extend beyond the greatest extent of the lesion to ensure complete removal of remaining satellite cysts or epithelial remnants of the cyst wall.

The primary difference between an en bloc osseous resection and other types of "peripheral" osseous management, such as curettage or ostectomy via a curette or bur, is the removal of the lesion in to with a surrounding margin of bone. Any possibility of violating the cyst capsule or leaving cyst remnants behind is greatly reduced. So the same principle is used here as it was a recurrent case which was treated earlier by enucleation and marginal resection. Osseous resection from lower left lateral incisor to angle of mandible and reconstruction was carried out with 5th rib graft taken from right side using 2.5 mm reconstruction plate. After 2 year of follow-up, there were no signs of recurrence and graft had taken up well.[13-17]

Peripheral ostectomy is primarily used as an adjunctive for osseous removal when resections can be avoided. Almost all theories of recurrence involve the possibility of leaving fragments of cyst lining or capsule within the bony cavity. One of the inherent problems with a peripheral ostectomy, just like curettage, is the "immeasurability" of the amount of osseous resection. Methylene blue can be used which penetrates bone when applied topically to a cavity to ensure proper clearance.[18]

Conclusions

Treating keratocystic odontogenic tumor (KCOT) has been one of the most controversial entities of the maxillofacial region. The priority of the treatment method chosen should depend on the size of the lesion, morbidity, patients' quality of life and recurrence rate. Even, large cysts can be successfully treated with the conservative approach. Regardless of the treatment option, both clinical and radiographic follow-up remains mandatory for years after surgery because recurrence may occur many years later.

References

1. Madras J, Lapointe H. Keratocystic odontogenic tumour: Reclassification of the odontogenic keratocyst from cyst to tumour. J Can Dent Assoc 2008;74:165-165h.
2. Philipsen HP. Keratocystic odontogenic tumour. In: Barnes L, Eveson JW, Reichart P, Sidransky D, editors. World Health Organization Classifi cation of Tumours. Pathology and Genetics of Head and Neck Tumours. Lyon: IARC; 2005. p. 306-7.
3. Shear M. The aggressive nature of the odontogenic keratocyst: Is it a benign cystic neoplasm? Part 1. Clinical and early experimental evidence of aggressive behaviour. Oral Oncol 2002;38:219-26.
4. Gomes CC, Gomez RS. Odontogenic keratocyst: A benign cystic neoplasm? Oral Oncol 2007;43:619-20.
5. Stoelinga PJ. Etiology and pathogenesis of keratocysts. Oral Maxillofac Surg Clin North Am 2003;15:317-24.
6. Pogrel MA, Jordan RC. Marsupialization as a definitive treatment for the odontogenic keratocyst. J Oral Maxillofac Surg 2004;62:651-5.
7. Morais de Melo W, Pereira-Santos D, Sonoda CK, Hochuli-Vieira E. Decompression for management of keratocystic odontogenic tumor in the mandible. J Craniofac Surg 2012;23:e639-40.
8. Shudou H, Sasaki M, Yamashiro T, Tsunomachi S, Takenoshita Y, Kubota Y, *et al*. Marsupialisation for keratocystic odontogenic tumours in the mandible: Longitudinal image analysis of tumour size using 3D visualised CT scans. Int J Oral Maxillofac Surg 2012;41:290-6.
9. Pitak-Arnnop P, Chaine A, Oprean N, Dhanuthai K, Bertrand JC, Bertolus C. Management of odontogenic keratocysts of the jaws: A ten-year experience with 120 consecutive lesions. J Craniomaxillofac Surg 2010;38:358-64.
10. Giuliani M, Grossi GB, Lajolo C, Bisceglia M, Herb KE. Conservative management of a large odontogenic keratocyst: Report of a case and review of the literature. J Oral Maxillofac Surg 2006;64:308-16.
11. Stoelinga PJ. The treatment of odontogenic keratocysts by excision of the overlying, attached mucosa, enucleation, and treatment of the bony defect with carnoy solution. J Oral Maxillofac Surg 2005;63:1662-6.
12. Zhao YF, Wei JX, Wang SP. Treatment of odontogenic keratocysts: A follow-up of 255 Chinese patients. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2002;94:151-6.
13. Mendes RA, Carvalho JF, van der Waal I. Characterization and management of the keratocystic odontogenic tumor in relation to its histopathological and biological features. Oral Oncol 2010;46:219-25.
14. Blanas N, Freund B, Schwartz M, Furst IM. Systematic review of the treatment and prognosis of the odontogenic keratocyst. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2000;90:553-8.
15. Stoelinga PJ. The management of aggressive cysts of the jaws. J Maxillofac Oral Surg 2012;11:2-12.
16. Jurisic M, Andric M, dos Santos JN, Jurisic V. Clinical, diagnostic and therapeutic features of keratocystic odontogenic tumors: A review. J BUON 2012;17:237-44.

17. Auluck A, Pai KM. Treatment of recurrent odontogenic keratocyst: A known but forgotten point. Br J Oral Maxillofac Surg 2006;44:74-5.
18. Abdullah A. Surgical treatment of keratocystic odontogenic tumour: A review article. Saudi Dent J 2011;23:61-5.

How to cite this article: ???

Source of Support: Nil, **Conflict of Interest:** None declared.